

Pilar cyst on the dorsum of hand A case report and review of literature

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Abstract

Rationale: Pilar cyst mainly occurs on the scalp, but pilar cyst on the dorsum of hand has not been reported. Herein, we provide information to improve the clinical cognition of pilar cyst location.

Patients concerns: A 76-year-old man presented with a round nodule on the opisthenar of his right hand for two months without any subjective symptoms.

Diagnoses: Histological features of the lesion biopsy indicated the diagnosis of pilar cyst.

Interventions: Surgical resection was made under local anesthesia.

Outcomes: Complete recovery was achieved after surgery.

Conclusion: Pilar cyst rarely occurs on the dorsum of hand and its diagnosis depends on histopathological examinations. Surgical resection is the only way to treat it.

Abbreviation: PLCD1 = phospholipase C delta 1.

Keywords: histological features, opisthenar, pilar cyst

1. Introduction

Pilar cysts are identified by Pinkus as the keratinization of the outer root sheath of hairs,^[1] which were originally called sebaceous cysts. The cysts are characterized by smooth, round nodules with solid texture and good mobility. It cannot be distinguished from epidermal cysts clinically, except that 90% of pilar cysts are found on the scalp where hair follicles are abundant. Other less common locations include face, trunk and extremities. Lesions rarely arise in palms, genitalia, axillary and groin.^[2] As far as we know, pilar cyst on the hand has rarely been reported before. The present study reports one such case on dorsum of hand in a male patient.

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This study was approved by the institutional review board of Xi'an Jiaotong University. Informed written consent was obtained from the patient for publication of this case report and accompanying images.

The datasets generated during and/or analyzed during the current study are publicly available.

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2. Case report

A 76-year-old man presented with a flesh-colored, dome-shaped nodule on the opisthenar of his right hand (Fig. 1), which had gradually increased in size for two months. The patient did not have any subjective symptoms. There is no history of trauma or chronic irritation at the site of the lesion and no similar lesion in his family members. He did not receive any treatment before he came to our hospital. The patient is generally in good condition. No changes in diet, sleep, urination, defecation, or body weight could be found. Physical examination showed that the patient possessed stable vital signs. The systematic examination revealed no evident abnormalities. A dermatological examination revealed a skin-colored nodule with smooth surface on dorsum of his right hand. This soybean-sized nodule was tough in texture with clear boundary and no hair follicle.

2.1. Auxiliary examination

There were no obvious abnormalities in blood routine, coagulation and liver and kidney function. A histopathological examination was conducted and the pathology showed that the cyst was in the reticular dermis (Fig. 2), with surrounding basal cells arranged as a fence. The cytoplasm of cells above the basal layer was lightly stained, swollen, and eosinophilic with fuzzy borders. The cells of stratified epithelium lost their nuclei abruptly without an intermediated granular cell layer. The innermost cells seemed to have fallen off into the cavity (Fig. 3).

2.2. Diagnosis

Clinically, it was considered as dermatofibroma at first. Based on histopathological examination, a diagnosis of pilar cyst was made.

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Figure 1. Appearance of the lesion on the back of right hand.

2.3. Therapeutic intervention and follow up

Complete excision under local anesthesia was done. After the treatment, the patient achieved complete recovery with no relapse during the 6 months of clinical follow-up.

3. Discussion

Pilar cyst is derived from external root sheath of the follicular isthmus, which is also referred to as isthmus-catagen cyst. Pilar cyst accounts for 20% of epithelial cysts and the others are epidermal.^[3] Compared with epidermal cysts, the lesions of pilar cyst do not have an overlying punctum and tend to be more mobile and firmed. Clinically, pilar cyst is indistinguishable from epidermal cyst, histopathological examination is indispensable to make a definite diagnosis. Pilar cysts are intradermal cysts with distinctive histological features. The cells of the cyst wall are composed of epithelial cells without obvious intercellular bridges. The inner cells suddenly transform into solid eosinophilic-staining keratin with no intervening granular layer. Nuclei of cells that fall into the cystic cavity generally disappear, but some cells still have nuclei. The cyst contents are homogeneous. Calcification exists in about 25% of cases. If the capsule wall is ruptured, a granulomatous reaction may occur. The cyst can therefore partially or completely disintegrate.^[4]

Pilar cyst often occurs in middle age with an obvious female preference and inheritance pattern of multiple cysts manifests as autosomal dominance.^[5,6] Hörer et al propose a monoallelic



Figure 2. Hematoxylin-eosin (H&E) staining of the lesion, showing a cyst in the reticular dermis and eosinophilic material. Bar length=6 mm.



Figure 3. No granular layer is observed at the wall of cyst (H&E). Bar length = $200 \,\mu\text{m}$.

mutation in phospholipase C delta 1 (PLCD1) that result in formation of multiple pilar cysts.^[7] Although pilar cyst is derived from follicular, several cases have indicated that pilar cyst can arise in non-hair bearing areas. The onset of pilar cyst was thought to be induced by inflammation and trauma.^[8–10] In addition, infection of the human papilloma virus may be related to the pathogenesis of pilar cyst.^[9] In very rare cases, pilar cyst can develop into malignant proliferative lump which clinically present as progressively enlarged lobulated masses, similar to squamous cell carcinoma.^[11] It is very important to distinguish pilar cysts from proliferating pilar cysts,^[12] since the later can undergo malignant transformation. Complete excision of the cyst is curative, but it is not recommended to remove surgically when the cyst is inflamed. Proliferating pilar cysts might need radiation therapy and/ or chemotherapy after surgical excision^[4].

The specificity of our case lies in its location. Compared to the incidence of scalp, pilar cyst on hand is extremely uncommon. To our knowledge, there are four cases of pilar cyst reported on hand previously, of which three are in finger tips and the other one is on the dorsum of the thumb. They were all proximal to the nail bed, which aroused the speculation that pilar cyst may be derived from nail matrix.^[13–16] The pilar cyst of our patient arose in the dorsum of right hand, a rare location and away from nail bed. No hair follicle was found. In other words, it can neither originate from hair follicles nor from nail matrix. Our case urges rethinking of the origin of pilar cyst.

Author contributions

ML and YF diagnosed and treated the patient. HH, YZ and SX conducted histopathological examination. ML and YF wrote the manuscript. All authors have approved the final article be true.

References

- Pinkus H. "Sebaceous cysts" are trichilemmal cysts. Arch Dermatol 1969;99:544–55.
- [2] Karaman E, Duman C, Yagiz C. Giant trichilemmal cyst at the neck region. J Craniofac Surg 2009;20:961–2.
- [3] Chandrasekaran V, Parkash S, Raghuveer CV. Epidermal cysts-a clinicopathological and biochemical study. Postgrad Med J 1980;56: 823–7.
- [4] Al Aboud DM, Patel BC. Pilar Cyst. Treasure Island, FL: StatPearls; 2019.

- [5] Poiares Baptista A, Garcia ESL, Born MC. Proliferating trichilemmal cyst. J Cutan Pathol 1983;10:178–87.
- [6] Leppard BJ, Sanderson KV. The natural history of trichilemmal cysts. Br J Dermatol 1976;94:379–90.
- [7] Shimomura Y, O'Shaughnessy R, Rajan N. PLCD1 and Pilar Cysts. J Invest Dermatol 2019;139:2075–7.
- [8] Perez LM, Bruce JW, Murrah VA. Trichilemmal cyst of the upper lip. Oral Surg Oral Med Oral Pathol Oral Radiol Endod 1997; 84:58–60.
- [9] D'Avila DG, Kanno DT, de Castilho da Silva D, et al. A proliferating trichilemmal cyst in the perianal region: a case report. Int J Surg Case Rep 2018;53:175–8.
- [10] Madan S, Joshi R. Trichilemmal cyst of the penis in a paediatric patient. Sultan Qaboos Univ Med J 2015;15:e129–32.

- [11] Lee SJ, Choi KH, Han JH, et al. Malignant proliferating trichilemmal tumor of the lower eyelid. Ophthalmic Plast Reconstr Surg 2005;21:349–52.
- [12] Kim UG, Kook DB, Kim TH, et al. Trichilemmal carcinoma from proliferating trichilemmal cyst on the posterior neck. Arch Craniofac Surg 2017;18:50–3.
- [13] Melikoglu C, Eren F, Keklik B, et al. Trichilemmal cyst of the third fingertip: a case report. Hand Surg 2014;19:131–3.
- [14] Ikegami T, Kameyama M, Orikasa H, et al. Trichilemmal cyst in the pulp of the index finger: a case report. Hand Surg 2003;8:253-5.
- [15] El Hassani Y, Beaulieu JY, Tschanz E, et al. Proliferating trichilemmal tumor of the pulp of a finger: case report and review of the literature. Chir Main 2013;32:117–9.
- [16] Maheshwari K, Hindocha S, Yousif A. Rare presentation of pilar cyst of the thumb. World J Plast Surg 2019;8:259–61.